Kaohsiung Journal of Medical Sciences (2013) 29, 582–583

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LETTER TO THE EDITOR

Adult intussusception with perforation caused by hemangioma in the distal ileum

To the Editor,

Although intestinal intussusception is a common condition in children, it is a rare entity in adults. Adult intussusception represents 5% of all cases of intussusception and accounts for only 1–5% of intestinal obstruction in adults [1]. Hemangiomas of the gastrointestinal tract are uncommon, accounting for only 0.05% of all gastrointestinal neoplasms [2]. Accordingly, intussusception caused by hemangioma is extremely rare, especially in adults. We report here a case of intussusception with perforation caused by hemangioma in the distal ileum in an adult. To the best of our knowledge, only one case of intussusception with perforation caused by hemangioma in an adult has been reported in the English-language literature [3].

A 64-year-old Chinese woman was admitted to our department with intermittent abdominal pain, accompanied by nausea, vomiting, and flatulence over a duration of 3 days. She denied any history of blood transfusion, alcohol abuse, or medication use. Her past clinical history and family history were normal. Physical examination showed a palpable mass measuring about 5 cm × 4 cm in the right lower abdomen with hyperactive bowel sounds (10–12 times/min).

Abdominal ultrasonography revealed a pseudo-kidney sign in the right lower quadrant, indicating intussusception (Fig. 1A). Magnetic resonance imaging was performed to verify a bulbiform mass with a bright signal intensity on T2-weighted images and an intermediate signal intensity on T1-weighted images, with a signal voided on T1- and T2-weighted images, at the ileocecal junction (Fig. 1B and C). Fat suppression was also revealed on T1- and T2-weighted images, indicating ileoileal intussusception (Fig. 1B and C). Colonoscopy revealed the mass to be a submucosal tumor at the ileocecal junction, with features of congestion (Fig. 1D).

On laparotomy, a reducible ileoileal intussusception was found in the distal ileum, lying 10 cm from the ileocecal junction. On reduction, a mass was palpable at the site of origin of the intussusception, and a perforation (2 cm in size) was found at the same site. Resection of the involved segment and end-to-end anastomosis were performed. Histologic examination showed the vascular walls to be thickened focally by adventitial fibrosis (Fig. 1E). According to the histology, a final diagnosis of cavernous hemangioma was confirmed. The patient's postoperative course was uneventful.

Hemangiomas are defined as masses of capillaries, blood-filled endothelial-lined spaces, or a combination of these. Hemangiomas can be divided into three categories: (1) the capillary hemangioma, described as a small tuft of submucosal capillaries that expand intraluminally and may develop into a stalk-like mass; (2) a category represented by mixed capillary and cavernous hemangiomas; and (3) the most common type, which is the cavernous hemangioma.

The cavernous type, as in the case we have reported here, is clinically evident, presenting with such symptoms as acute or chronic gastrointestinal hemorrhage, anemia, and obstruction [4]. However, intussusception caused by hemangioma is extremely rare. Donhauser and Kelly [5] found jejunal hemangiomas to be responsible for only one of their 665 cases of intussusception. Very few cases have been described in the English-language literature, as shown by searching for "hemangioma," "intussusception," and "adult" on the Medline database. Accordingly, adult intussusception with perforation caused by hemangioma is still rarer. To the best of our knowledge, only one case has been reported in the English-language literature [3].

In conclusion, we present a unique case of an intussusception with perforation caused by hemangioma in the distal ileum in an adult. It should be kept in mind that if there is an abdominal mass with intussusception and perforation, the possibility of hemangioma should be considered.

http://dx.doi.org/10.1016/j.kjms.2013.03.005

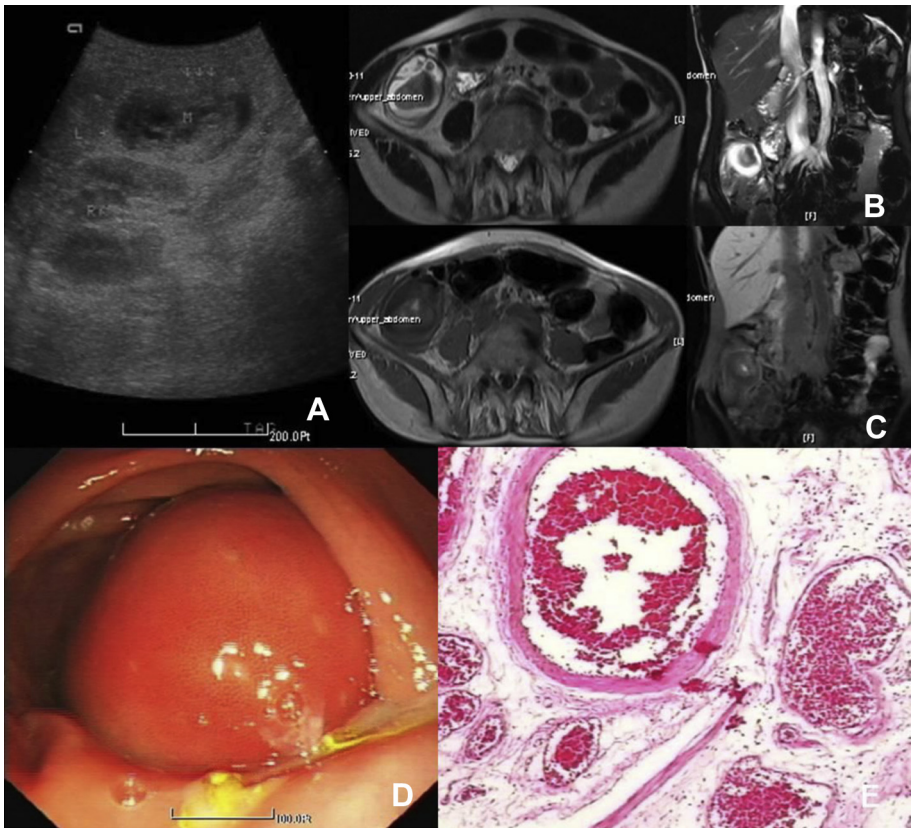

Figure 1. Abdominal ultrasonography revealed a pseudo-kidney sign at the right lower quadrant of the abdomen (A). Magnetic resonance imaging was performed to verify a bulbiform mass with a bright signal intensity on T2-weighted images and an intermediate signal intensity on T1-weighted images, with a signal voided on T1- and T2-weighted images, at the ileocecal junction (B, C). Fat suppression was also revealed on T1- and T2-weighted images (B, C). Colonoscopy revealed the mass to be a submucosal tumor at the ileocecal junction, with features of congestion (D). Histologic examination showed the vascular walls to be thickened focally by adventitial fibrosis (E; H&E stain, 100×).

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Ying Huang
Qiong Zhang
Nursing Department, Zhejiang Cancer Hospital, Hangzhou, China

Ji-Feng Feng*
Department of Oncological Surgery, Zhejiang Cancer Hospital, Hangzhou, China

Huang Liu
Department of Clinical Medicine, Medical College of Xiamen University, Xiamen, China

Jing Liu
Department of Clinical Medicine, Medical College of Xiamen University, Xiamen, China

Department of General Surgery, Southeast Hospital of Xiamen University, Zhangzhou, China

*Corresponding author. Department of Oncological Surgery, Zhejiang Cancer Hospital, 38 Guangji Road, Hangzhou 310022, China.
E-mail address: Jifzhejiang@gmail.com